<Case Report>Intussusception caused by primary malignant melanoma of the small intestine

AUTHOR(S):

KOGIRE, MASAFUMI; YANAGIBASHI, KEN; SHIMOGOU, TSUKASA; IZUMI, FUYUKI; SUGIYAMA, AKIO; IDA, JUN; MORI, AKIRA; ... OGAWA, HIROKI; SAIGA, TAKAYOSHI; SAKANASHI, SHIRO

CITATION:

KOGIRE, MASAFUMI ...[et al]. <Case Report>Intussusception caused by primary malignant melanoma of the small intestine. 日本外科宝函 1996, 65(2): 54-58

ISSUE DATE:

1996-05-01

URL:

http://hdl.handle.net/2433/203558

Arch Jpn Chir 65(2), 54～58, Mai., 1996

Intussusception caused by primary malignant melanoma of the small intestine

Masafumi Kogire, Ken Yanagibashi, Tsukasa Shimogou, Fuyuki Izumi, Akio Sugiyama, Jun Ida, Akira Mori, Jun Tamura, Nobuo Baba, Hiroki Ogawa, Takayoshi Saiga* and Shiro Sakanashi

Department of Surgery and *Pathology, Otsu Red-Cross Hospital, Otsu, Shiga, Japan
Received for publication, Apr. 1, 1996

Abstract

Whether melanoma develops as a primary tumor in the small bowel remains controversial. A 57-year-old male Japanese presented signs of intestinal obstruction. Ultrasonography and computed tomography disclosed an abdominal mass with multiple concentric rings, characteristic of intussusception. At surgery, a spherical tumor, 3.8 cm in diameter, with scattered pigmentation was found to lead the intussusception. Segmental intestinal resection with regional lymph node dissection was performed. Pathological examination revealed diffuse infiltration of malignant melanoma cells. Nodal metastasis was seen only in the mesenteric node draining from the tumor-bearing intestinal segment. Twelve months after surgery, melanoma recurred in the liver and para-aortic lymph nodes, where a malignancy of the digestive organs frequently metastasizes; however, no extraperitoneal melanoma was found after repeated examinations. Thus, this case suggests that primary malignant melanoma can originate in the small intestine and be a cause of intussusception in the adults.

Introduction

Adult intussusception is uncommon, and associated with a neoplasm in half of cases (1). Among the causative neoplasms is intestinal melanoma, which is considered exclusively metastatic (2). Whether melanoma develops as a primary tumor in the small bowel remains controversial (3). We report a case of intussusception caused by primary malignant melanoma of the small intestine.

Case report

A 57-year-old male Japanese presented with a 2-day history of abdominal pain and vomiting. A mobile mass measuring 5 × 5 cm was palpable to the right of the umbilicus. Laboratory examination showed moderate anemia (hematocrit 27.9%). Plain abdominal radiography showed air-fluid levels in dilated bowel loops. Ultrasonography (Fig. 1) and computed tomography (Fig. 2) disclosed

Present Address: Department of Surgery & Surgical Basic Science, Graduate School of Medicine, Kyoto University, 54-Shogoin Kawara-cho, Sakyo-ku, Kyoto 601-01, Japan
索引用語: 腸重積, 小腸, 黒色腫
Key words: Intussusception, small intestine, melanoma

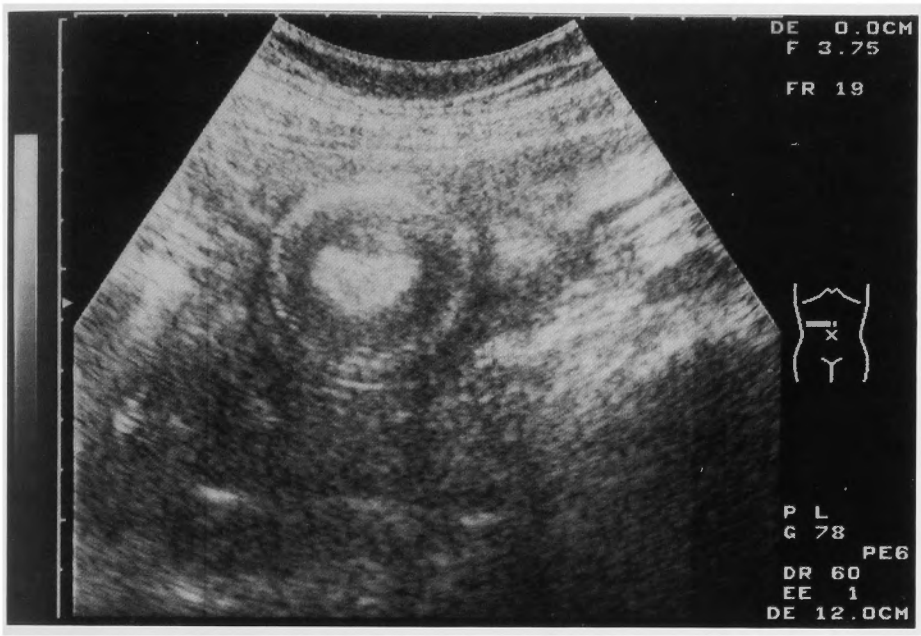

Fig. 1 Ultrasonogram showing a mass with hyperechoic and hypoechoic concentric rings, characteristic of intussusception.

Fig. 2 Computed tomogram showing a mass with enhanced and non-enhanced concentric rings, characteristic of intussusception.

an abdominal mass with multiple concentric rings, characteristic of intussusception (4). At surgery, an intussusception 100 cm from the ligament of Treitz was found. Segmental resection with regional lymph node dissection was performed. A spherical, firm tumor, 3.8 cm in diameter, with scattered pigmentation led the intussusception (Fig. 3). Microscopic examination showed diffuse infiltration of the tumor cells with brown pigments (Fig. 4). S100 protein, a diagnostic indicator of ma-

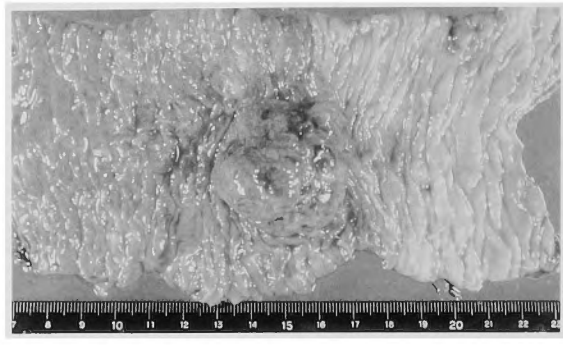

Fig. 3 Resected segment of the small intestine with a spherical, firm tumor showing scattered pigmentation.

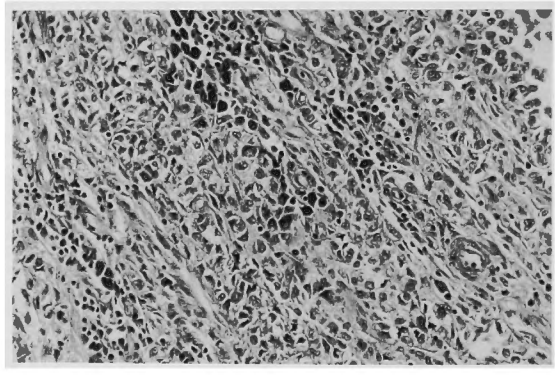

Fig. 4 Histopathological section showing diffuse infiltration of the tumor cells with brown pigments (Hematoxylin and eosin, ×100).

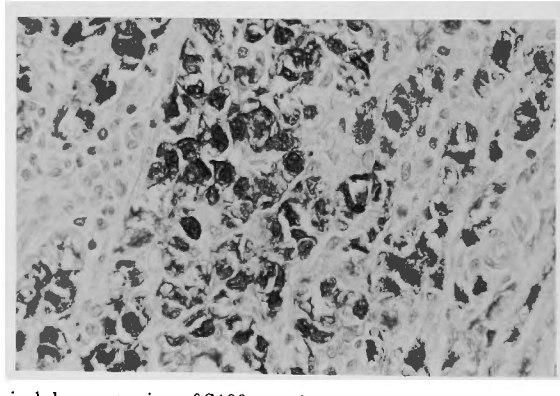

Fig. 5 Immunohistochemical demonstration of S100 protein, a diagnostic indicator for malignant melanoma, in the tumor cells (×400).

lignant melanoma (5), was demonstrated immunohistochemically in the tumor cells (Fig. 5). Metastasis was microscopically confirmed in one of the mesenteric lymph nodes. No cutaneous, mucocutaneous, ocular or nasopharyngeal melanoma was found postoperatively after repeated thorough examinations. Twelve months after surgery, metastases to the liver and to the para-aortic lymph nodes appeared. The patient died from progression of liver metastases 21 months after surgery.

Discussion

Although preoperative diagnosis of adult intussusception is often difficult to make (1, 4), our case showed signs strongly suggestive of intussusception: clinical symptoms and radiological evidence of intestinal obstruction, and an abdominal mass. A multiple concentric ring sign demonstrated by ultrasonography and computed tomography is a characteristic feature of intussusception (4).

In contrast to childhood intussusception, which is idiopathic in 90% of cases, adult intussusception is associated with a causative lesion in 75 to 90% of cases (1, 4). Neoplasms account for about 50% of adult intussusception (1). Melanoma of the small intestine can cause intussusception (1, 2). Because melanoma frequently metastasizes to the small bowel (2) and because intestinal mucosa usually contains no melanoblasts, melanoma of the small intestine is thought to be metastatic even if no other primary site was detected (6). However, some authors have proposed the existence of primary malignant melanoma of the small intestine (3, 7).

In our case, several lines of evidence indicate that melanoma developed as a primary tumor in the small intestine. First, nodal metastasis was seen at surgery only in the mesenteric node draining from the tumor-bearing intestinal segment. This illustrates the typical spread of melanoma from a primary site to regional lymph nodes. Second, our patient lived for 21 months after surgery without evidence of extraperitoneal metastases. In contrast, most patients in whom intraabdominal metastases were the first manifestation of the disease exhibited evidence of extraperitoneal involvement during the course of their disease and died within one year (6). Third, melanoma recurred in our patient in the liver and para-aortic lymph nodes, where a malignancy of the digestive organs frequently metastasizes. Thus, this case suggests that primary malignant melanoma can originate in the small intestine and be a cause of intussusception in the adults.

Melanoblasts are thought to originate from neural crest cells (8). In the gut, neural crest cells become amine precursor uptake and decarboxylation (APUD) cells (7). Therefore, it is possible that melanoma of the intestine originates from APUD cells as suggested by *Krausz et al.* (7). The findings that S100 protein, which has been shown in most cases of malignant melanoma including our case, is distributed in many kinds of neural crest derivatives (5) are in accordance with this concept.

References

1) Nagorney DM, Sarr MG, McIlrath DC: Surgical management of intussusception in the adult. Ann Surg **193**: 230–236, 1981.
2) Klaase JM, Kroon BBR: Surgery for melanoma metastatic to the gastrointestinal tract. Br J Surg **77**: 60–61, 1990.
3) Kadivar TF, Vanek VW, Krishnan EU: Primary malignant melanoma of the small bowel: a case study. Am Surg **58**: 418–422, 1992.
4) Reijnen HAM, Joosten HJM, de Boer HHM: Diagnosis and treatment of adult intussusception. Am J Surg

158: 25–28, 1989.
5) Nakajima T, Watanabe S, Sato Y, et al: Immunohistochemical demonstration of S100 protein in malignant melanoma and pigmented nevus, and its diagnostic application. Cancer 50: 912–918, 1982.
6) Baab GH, McBride CM: Malignant melanoma. The patient with an unknown site of primary origin. Arch Surg 110: 896–900, 1975.
7) Krausz MM, Ariel I, Behar AJ: Primary malignant melanoma of the small intestine and the APUD cell concept. J Surg Oncol 10: 283–288, 1978.
8) Rosai J: Skin/tumors and tumorlike conditions. In Ackerman's Surgical Pathology, 6th ed., edited by Rosai J, St. Louis, The C V Mosby 1981, pp 81–152.

和文抄録

腸重積にて発症した小腸悪性黒色腫の1例

大津赤十字病院 外科
小切 匡史，柳橋 健，下郷 司，泉 冬樹
杉山 昌生，井田 純，森 章，田村 淳
馬場 信雄，小川 博暉，坂梨 四郎

大津赤十字病院 病理
雑賀 興慶

非常に稀である，腸重積にて発症した小腸原発の悪性黒色腫の1例を報告する．症例は57歳の男性で，腹痛と嘔吐を主訴に来院し緊急入院となった．入院時，臍の右方に5×5 cmの可動性のある腫瘤を触知し，検査所見では中等度の貧血があり，立位腹部単純レ線で小腸ニボーを認めた．造影CT及びUSでは臍の右方に腸重積に典型的な層構造を示す腫瘤像を認めた．小腸重積によるイレウスと診断し手術を行ったところ，トライツ靭帯より1m の空腸に所々に黒色を呈する部分のある3.8×3.8 cm の1型の腫瘤があり，それを先進部として腸重積をきたしていた．腸重積を解除し，空腸を約20 cm切除し，手術を終えた．切除標本の組織像では茶褐色の色素沈着を伴う腫瘍細胞の増殖を認め，悪性黒色腫に一致した所見であり，腸間膜リンパ節に1個転移を認めた．術後12ヶ月目に肝転移と大動脈周囲リンパ節転移が出現し，術後21ヶ月目に肝不全のため死亡したが，腹腔外には度重なる検査にも関わらず原発巣と考えられる病変は指摘できなかった．

悪性黒色腫の原発部位は皮膚，皮膚粘膜移行部，眼などであり，小腸は本疾患の転移の好発部位のひとつであるが，小腸原発例は非常に稀である．我々の症例は，小腸病巣の所属リンパ節に転移があったこと，術後の詳細な検索にもかかわらず，皮膚その他に原発巣と考えられる病変を認めなかったこと，再発部位（肝，大動脈周囲リンパ節）が腹腔内臓器由来の悪性疾患の再発好発部位に一致したことより，悪性黒色腫が小腸に原発する可能性があり，さらにそれが成人における腸重積の原因となりうることを示唆していると考えられた．